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Primary splenic hydatid cyst treated with laparoscopic spleen preserving surgery: A case report

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ABSTRACT

BACKGROUND: Primary splenic hydatidosis is an uncommon etiology. Total splenectomy has been the treatment of choice, however, spleen preserving surgery has been gaining popularity.

CASE REPORT: We present a case of a 52-year-old man who was evaluated for a splenic cyst, found incidentally on CT scan done at another institution. MRI was repeated in our institution along with an indirect hemagglutination test to confirm the diagnosis. Since our patient had a single non-complicated cyst confined to the spleen's lower pole, we decided to opt for laparoscopic cyst unroofing and omentoplasty, a spleen preserving technique.

DISCUSSION: Due to the rarity of splenic hydatidosis, no strict management guidelines are available, and the decision is usually left for the surgeon. Total splenectomy has been considered the standard of care, however, spleen preserving surgical techniques and percutaneous interventions have been suggested as better alternatives. Spleen preserving techniques lead to surgical outcomes comparable to total splenectomy, with the added benefit of preserving the immunological protection provided by the spleen and protecting patients from dramatic complications that might follow total splenectomy, such as overwhelming post-splenectomy infections, which are associated with very high mortality.

CONCLUSION: Laparoscopic cyst unroofing and omentoplasty is a feasible surgical technique to treat primary splenic hydatidosis while preserving the spleen's immunologic function.

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1. Introduction

Primary splenic hydatid cysts are a rare entity representing less than 3.5% of total abdominal hydatidosis [1,2]. Splenectomy is considered the standard of care when it comes to splenic hydatidosis, however, this procedure is associated with an increased risk of overwhelming post-splenectomy infection (OPSI), which can be fatal in many patients. For this reason, spleen preserving techniques, which include laparoscopic or open partial splenectomy and percutaneous drainage, have been emerging as an alternative. These techniques provide comparable outcomes in terms of recurrence, and preserve the immunological protection provided by the spleen. We report a case of a man whom we treated for pri-

mary splenic hydatidosis by laparoscopic partial splenectomy with omentoplasty.

This case was reported in line with the SCARE criteria [3].

2. Case presentation

A 52-year-old, previously healthy man, residing in China for the past nine years, was seen in the emergency department for dry cough, fever, and night sweats occurring for one week. A chest X-ray and Computed Tomography (CT) scan of the chest were ordered as part of the workup for a possible respiratory infection. The chest x-ray showed no visible lung or mediastinal pathologies, however, an abnormal lucency in the left upper quadrant of his abdomen was noted. The CT confirmed the absence of thoracic pathologies, however, lower cuts through the upper abdomen showed splenomegaly, with a cystic mass at the lower pole of the spleen and an intraluminal floating membrane suggestive of a hydatid cyst (Fig. 1).

An abdominal ultrasound was then done, as part of the workup for the splenic mass. It showed an enlarged spleen, containing a thin-walled 14.2 cm × 9.9 cm septated cyst near the lower edge of

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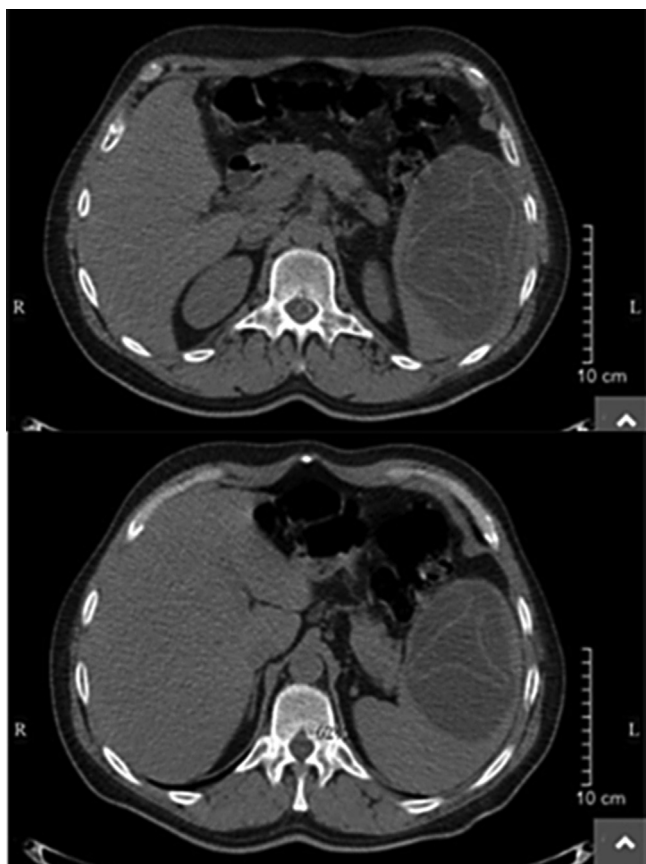


Fig. 1. Axial images of a non-enhanced CT scan of the abdomen showing a large 14.5 cm well defined hypodense cystic mass of spleen with internal thick dense septations.



Fig. 2. Longitudinal grey-scale images of the spleen which show a 14.2 cm anechoic splenic cyst that shows enhanced transmission through, and contains thick echogenic undulating septations.

the organ (Fig. 2). No additional cystic lesions were identified in the liver or anywhere else in the abdomen.

The patient presented to our center a week later. He reported a recurrent occasional dull aching pain in his left subcostal region occurring for a few months. The physical exam did not reveal any findings of interest except for a palpable spleen. He provided the imaging and laboratory results he did in China. As for all patients referred to us from other institutions, imaging and laboratory investigations were repeated at our center, as per our hospital's protocol. We decided to opt for an abdominal MRI, in addition to routine labs, and an indirect hemagglutination test.

Labs showed slight eosinophilia (4.6%), and the indirect hemagglutination test was positive (1/256). The abdominal MRI showed

an enlarged spleen containing a well-defined capsulated 14.5 cm cyst at the lower edge, with an intraluminal floating membrane representing the “Water Lilly” sign, compatible with a large hydatid cyst (Fig. 3). The rest of the examination was normal and no cysts were identified in the liver. A final diagnosis of primary splenic hydatid cyst (SHC) was done. The patient was scheduled for laparoscopic cyst unroofing with omentoplasty, a spleen preserving technique. Albendazole was started 3 days before the operation.

Laparoscopic exploration revealed a huge cyst at the lower splenic border. A cetrimide-soaked gauze was placed around the spleen as a precautionary measure. The cyst was injected and irrigated with cetrimide 0.5% solution, which was allowed 10 min to act. After that, the cyst was punctured, and 300 mL of crystal clear fluid were aspirated. A white membrane was observed collapsed inside the cystic lumen and was excised en bloc. Before closing, a flap of the omentum was moved to fill the residual cavity in the spleen (Fig. 4). The operation time was 2 h and 54 min, and the excised specimen was sent to pathology.

The patient tolerated the surgery well and was sent home on the third postoperative day. He was prescribed albendazole for 28 days, and we recommended labs and imaging to be repeated in 4 months. Later the same week the pathology report confirmed our diagnosis of a splenic hydatid cyst (Fig. 5). No signs of recurrence were found after 4 months during follow up CT scan.

3. Discussion

Hydatid disease is caused by a human infestation by the larvae of one of the Echinococcal species, most commonly echinococcus granulosus [4]. The disease usually involves the liver (75%), the lungs (15.4%), and the spleen (5.1%), and is the top cause of splenic cysts in endemic areas [5]. First identified by Berlot 1791 during a cadaveric autopsy [6], splenic hydatid cysts account for 0.5%–8% of all hydatidosis [2]. Isolated involvement of the spleen, however, is rare, representing only 1.5%–3.5% of abdominal hydatidosis according to several case series [1,2]. An infestation of the spleen can happen by one of three routes. The first, the arterial route, happens after the organism passes through the liver and the lung, and ends up in the spleen. The second is the venous route, through the portal circulation, avoiding passage through the liver. A third route is through secondary infestation of the spleen after systemic dissemination or intraperitoneal seeding following the rupture of a hepatic cyst [7]. Our patient had primary splenic hydatidosis, and therefore, the first or the second routes could apply to his situation.

Hydatid cysts usually grow at a rate of 0.3 cm–1 cm per year [5], and hence symptoms may take several years before starting to appear. In fact, in up to 30% of cases, splenic hydatid cysts are an incidental finding, like our case [8]. When symptomatic, patients mostly complain of left upper quadrant pain, or symptoms related to the mass effect by the enlarged spleen [9].

The diagnosis of splenic hydatid cysts can be challenging for physicians. Imaging studies with ultrasound, computed tomography, or magnetic resonance have all shown comparable success in achieving the diagnosis [5]. Serological tests such as ELISA, immunoelectrophoresis, or indirect Hemagglutination test can also assist in the diagnosis [5]. In our case, the patient interestingly underwent imaging by the three different imaging modalities mentioned. This gave us the chance to view the same cyst in 3 different ways pre-operatively and establish the diagnosis. From the serological tests, we chose the indirect Hemagglutination test for its availability.

Since it's a rare entity, there are no strict recommendations regarding the treatment of splenic hydatid cysts. Surgical treatment has been the standard of care in splenic hydatid cyst disease, with total splenectomy being the conventional treatment in previous years [10]. Laparoscopic surgery has shown better results in

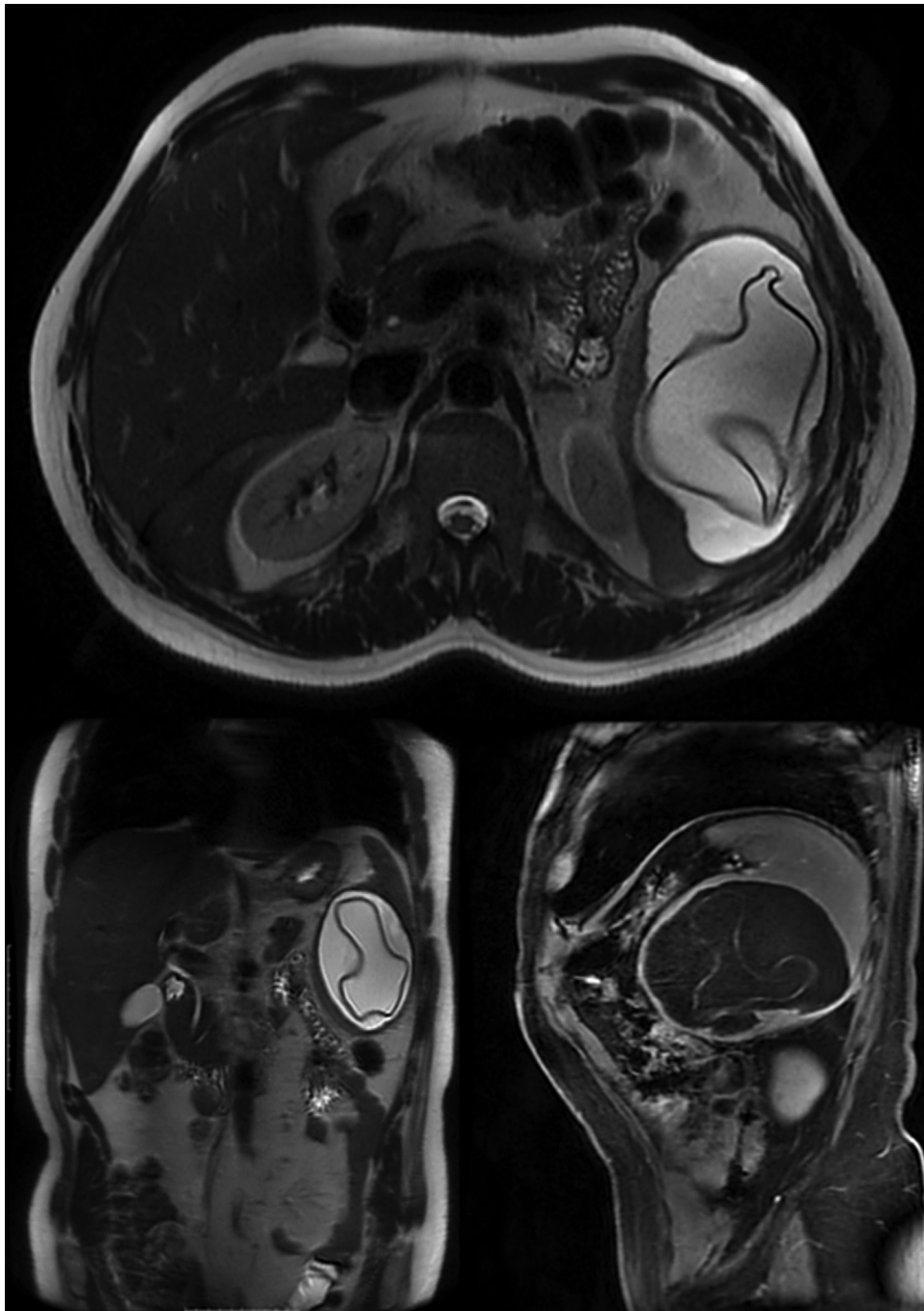


Fig. 3. (A) and (B) axial and coronal T2 weighted images (T2 single shot fast spin echo SSFSE) of 3 Tesla MRI that show a cystic lesion of the spleen. The cyst appears of high signal intensity with a hypointense rim (pericyst) and intraluminal septations mostly representing the detached membranes. Findings represent the “water lily sign”. (C) Sagittal T1 3D gradient echo fat sat post gadolinium injection images of the splenic cyst during the portal venous phase that show no enhancement of the cyst wall or septa.

previous studies, with significantly lower morbidity rates when compared to open technique [11]. Total splenectomy could lead to some dreadful comorbidities, most importantly an increased risk of overwhelming post-splenectomy infections (OPSI), which carries a mortality rate reaching 50% [12]. This has led to the rationale behind spleen preserving surgery, suggesting it as a better alternative to total splenectomy when feasible, since it could preserve the spleen’s immunological role and reduce such risks. Spleen preserving surgery includes cyst unroofing, partial cystectomy, partial splenectomy, and cystojejunal Roux-en-Y anastomosis [13,14]. These techniques have been suggested as a good option for cases with simple, single, small, and peripherally located cysts, especially in young patients [14]. Spleen preserving surgery does not appear

to be significantly associated with a higher recurrence rate when compared to total splenectomy [13], however, it can be more technically demanding and might carry a higher risk of bleeding due to the larger splenic remnant [5].

In recent years, PAIR technique, which stands for Percutaneous Aspiration Irrigation and Re-aspiration under radiographic guidance, has been gaining popularity in the treatment of splenic hydatid cysts. This method has shown results comparable to those gained with the surgical approach in terms of cure and relapse [15]. PAIR could be useful in treating small (<5 cm), Type I or II splenic hydatid cysts in patients who have high surgical risk or refuse surgery [5].

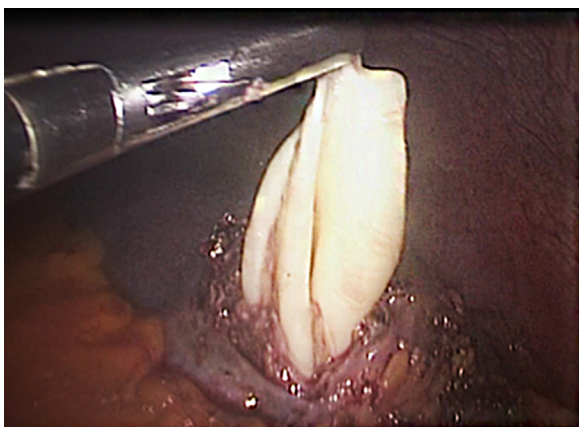


Fig. 4. Hydatid cyst membrane as seen during laparoscopy.

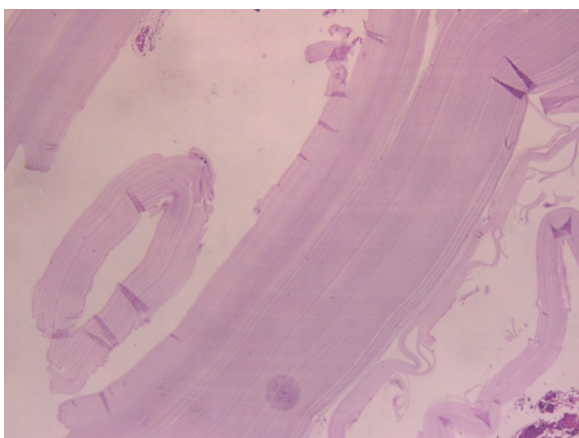


Fig. 5. Laminated membranes of a hydatid cyst involving the spleen (H&E stain, enlarged $\times 40$).

Splenic hydatidosis is an uncommon entity, with no strict recommendations guiding the treatment choice. Surgery and percutaneous intervention are the most commonly used modalities. Total splenectomy can carry a high rate of morbidity, therefore, spleen preserving techniques have been suggested as a suitable alternative. We believe that laparoscopic cyst unroofing is a feasible technique achieving results comparable to those seen with conventional splenectomy, while preserving the immunologic function of the spleen.

Declaration of Competing Interest

This article has no conflict of interest with any parties.

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Ethical approval

The study type is exempt from ethical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

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Provenance and peer review

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